American Journal of Men's Health
July-August 1–6

DOI: 10.1177/1557988320923913
journals.sagepub.com/home/jmh
SAGE

Superselective Arterial Embolization of Arteriovenous Malformation of Internal Pudendal Artery, a Rare Cause of Hematuria: A Case Report

Shixiang Qiu[1], Xin Wei[2], Hong Hu[2], Xiaowei Zhao[2], Xiaoxia Wen[2], and Li-Ming Zhong[2]

Abstract

Arteriovenous malformation (AVM) is a common form of vascular malformation, an abnormal direct communication between an artery and a vein without passing through the capillaries. AVM may just present as hematuria without plain or other symptoms. The article presents a case of a 52-year-old male with gross hematuria diagnosed as AVM of internal pudendal artery, which was successfully managed with superselective arterial embolization using temporary embolization materials.

Keywords

hematuria, arteriovenous malformation, TAE, interventional therapy

Received February 3, 2020; revised March 27, 2020; accepted April 9, 2020

Background

Hematuria is defined as the presence of red blood cells in the urine and is a problem commonly encountered by primary care physicians. Hematuria is the most common symptom of urinary system diseases (Bolenz et al., 2018; Davis et al., 2012), categorized as gross hematuria and microhematuria. Gross hematuria is less common than microscopic hematuria, but is more often associated with dangerous causes. Thus, the clinician should turn his or her attention to the patient with gross hematuria. Many diseases may cause hematuria, such as malignancy, benign tumor, infection, menstruation, urolithiasis, trauma, urinary anatomic disease, renal disease, chemical, and hematologic/vascular disease (Joshua et al., 2017; Peterson & Reed, 2019). The commonest cause is infection of the lower urinary tract, particularly infection of the bladder (Bin_ Dahman & Aljabry, 2019). According to a report, about 98% of hematuria is caused by diseases of the urinary system itself and only 2% caused by diseases of the whole body or adjacent organs of the urinary system (Linder et al., 2018). Malignancy is the commonest reason for gross hematuria, such as renal cancer, bladder cancer, and prostate cancer. Arteriovenous malformation (AVM) is a rare cause of gross hematuria.

AVMs are abnormal formations of blood vessels that shunt arterial blood directly into veins without passing through the capillaries. These vessels usually are crooked, dilated, and with thick vessel walls (Uller et al., 2014). The exact etiopathology of AVMs is unclear. The lack of blood flow and oxygen in the capillaries can lead to tissue damage in the affected areas. AVMs usually lead to the formation of a dilated sac-like connection, arteriovenous aneurysm, and so forth. Particularly, arteriovenous aneurysm is a fatal lesion. AVMs most commonly affect the head and neck (47.4%), followed by the extremities (28.5%). Extracranial AVMs are focal and diffuse. However an AVM that is associated with the internal pudendal artery is uncommon

[1]School of Medical Imaging, North Sichuan Medical College, Nanchong, Sichuan Province, P.R. China
[2]Department of Interventional Radiology, Central Hospital of Nanchong, The Second Clinical School of North Sichuan Medical College, Nanchong, Sichuan Province, P.R. China

Corresponding Author:
Li-Ming Zhong, Professor, Chief Physician, Department of Interventional Radiology, Central Hospital of Nanchong, The Second Clinical School of North Sichuan Medical College, 97 Renmin South Road, Central Hospital of Nanchong, Nanchong, Sichuan Province 637000, P.R. China.
Email: 625826874@qq.com

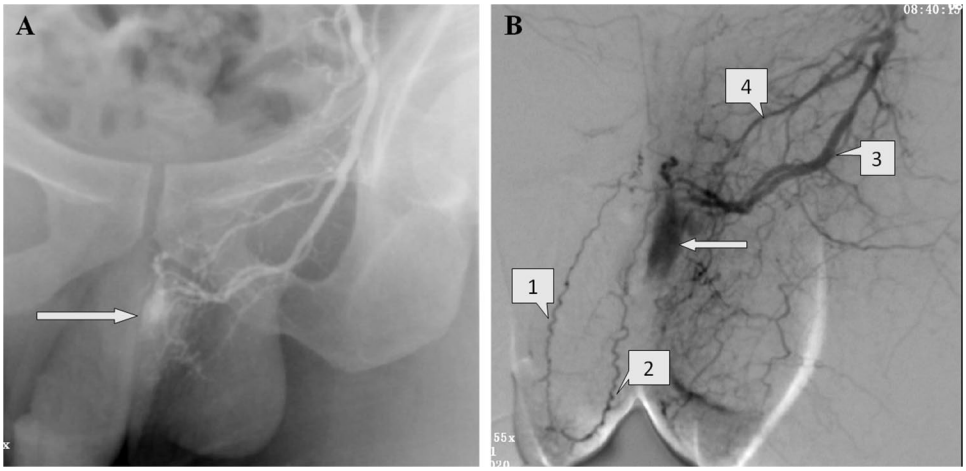

Figure 1. Imaging of digital subtraction angiography (DSA) demonstrates the existence of left arteriovenous malformation (AVM). AVM lesion is the white arrow shown in A and B; dorsal artery of penis; 2: deep artery of the penis; 3: left internal pudendal artery; 4: left prostatic arteries.

(Celtikci et al., 2014; Jesinger et al., 2013). Surgical treatment is complex due to location of lesions and endovascular approach is usually the first choice among the treatment options, such as superselective vascular embolization (Jesinger et al., 2013; Papadakos et al., 2008). The article presents a case report of AVM of the internal pudendal artery, which was successfully treated by transarterial embolization with gelatin sponge and pieces of silk line.

Case Presentation

A 52-year-old male visited a doctor because of a history of repeated episodes of unexplained painless gross hematuria in the past two decades. During the past 20 years, he had visited a local hospital four times for massive hematuria, which had alleviated with conservative medical treatment. This episode was so violent that there was no time for cystoscopy. He denied any other accompanying symptoms including fever, headache, periorbital edema, joint pain, abdominal or flank pain, weight loss, skin rash, bleeding tendency, dysuria, or intermittent micturition. Hematuria was not preceded by upper respiratory tract infection or related to urinary trauma or exercises. No history of blood transfusion or other special diseases existed. Urine analysis (UA), coagulation function test, and kidney function tests (KFTs) were repeatedly normal.

Further examinations were preformed to investigate the cause of hematuria. The measurements, echogenicity, and density of kidney were normal when investigated by computed tomography (CT) urography and urinary ultrasonography (US), which excluded the hydronephrosis, masses, or stones of the urinary tract. To exclude hematuria caused by vascular lesions,

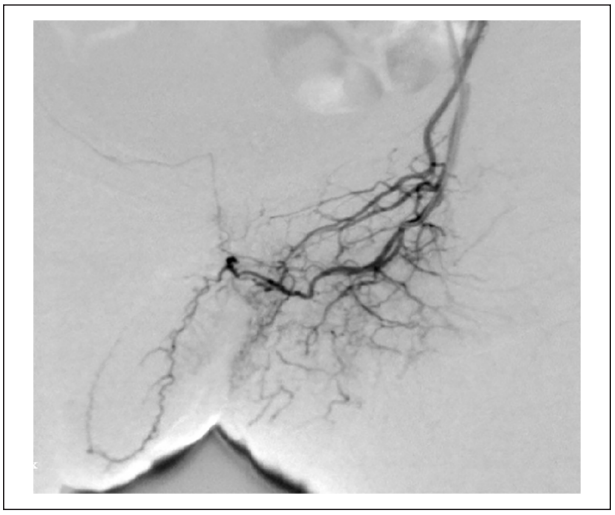

Figure 2. Imaging of superselective embolization of left internal pudendal artery. Arteriovenous malformation (AVM) lesion disappeared without extravasation of contrast medium outside the vascular spaces.

the patient underwent selective transfemoral pudendal arteriography. The arteriography was performed under local anesthesia via the right femoral artery with a 5 French sheath. Both internal iliac arteries were selectively catheterized, and there were no abnormal findings after contrast agent injection in both internal iliac arteries. However, the left pudendal artery was catheterized with a 1.7 French microcatheter and abnormal staining and extravasation of contrast agent of distal vascular branches was found, which confirmed the existence of AVM (Figure 1). Superselective embolization of the left

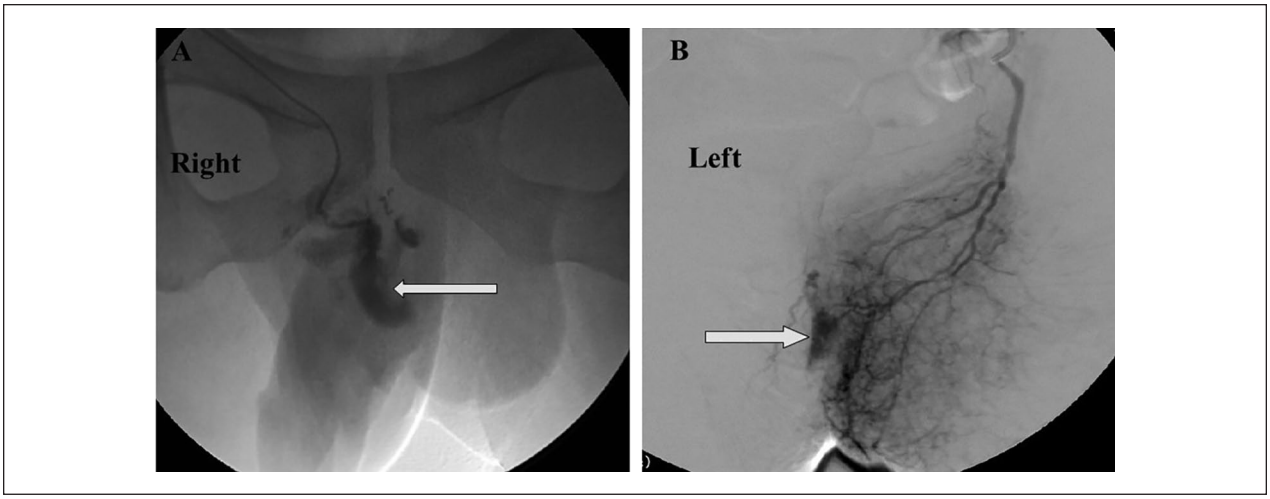

Figure 3. Imaging of digital subtraction angiography (DSA) demonstrates the existence of arteriovenous malformation (AVM). AVM lesion is the white arrow shown in both A and B. A: the right; B: the left.

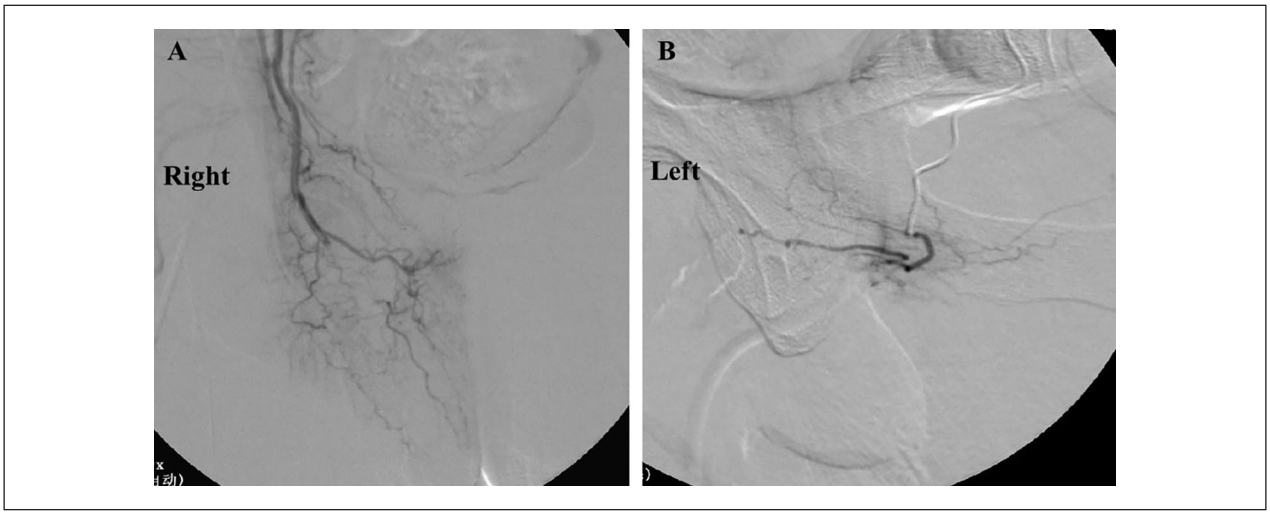

Figure 4. Imaging of superselective embolization of both internal pudendal arteries. Arteriovenous malformation (AVM) lesion disappeared without extravasation of contrast medium outside the vascular spaces. A: the right; B: the left.

internal pudendal artery was performed with infusion of gelatin sponge until complete embolization of the lesion. After embolization, control angiograms showed no evidence of AVM or contrast extravasation while the perineal blood flow remained sufficient (Figure 2). Hematuria disappeared and the color of urine became clear within 6 hours.

The hematuria recurred without any accompanying symptoms 2 years later. A digital subtraction angiography was performed again, and abnormal staining and contrast agent extravasation of the both internal pudendal artery distal vascular branches were found (Figure 3). The patient chose transcatheter embolization (TAE) as he did 2 years ago. Superselective embolization of the bilateral pudendal arteries was executed with infusion of gelatin sponge plus natural silk segments. No existence of extravasation of contrast medium outside the vascular spaces was found (Figure 4). The difference from the first time was the use of natural silk segments and the slow speed of embolization. After long-term follow-up of 10 years, no recurrence and no postoperative complications such as erectile dysfunction and ischemic necrosis were found.

Discussion

Hematuria is often a common symptom of urinary system disease. A good medical history, physical examination, and focused laboratory investigations for persistent or

recurrent gross hematuria may lead to the identification of a long list of causes including infection, glomerulonephritis, stones, trauma, tumor, and so forth (Joshua et al., 2017; Peterson & Reed, 2019). But many rare causes still exist, including AVM, vascular fistula, renal nutcracker syndrome, vesical varices, pyelovenous fistula, and so forth (Bin_ Dahman & Aljabry, 2019; Valdez Vargas et al., 2018). Some rare causes of gross hematuria require more detailed workup. This is true theoretically, but in reality many patients and doctors tend to ignore hematuria, especially asymptomatic hematuria, and attribute it for physiological or other unimportant reasons. As a cohort study reported, only 18% of patients in their study received a complete evaluation based on the American Urological Association guidelines, with 54% having no imaging or procedure performed (Ark et al., 2017).

Most guidelines recommend similar examination steps for gross hematuria (Schmitz-Drager et al., 2016). The first step is a good medical history taking to exclude causes that are unnecessary to treat, such as (previous) urinary tract infection, menstruation, strenuous sport activity, or medical intervention in the urinary tract. Patients with a history of any of these are excluded from further investigation. Then, the indicators of a nephrological cause are considered, usually by testing for albuminuria, sediment testing to assess red blood cell morphology, and measuring blood pressure and renal function. If no sign of glomerular kidney disease is found, further urological investigation with imaging of the upper urinary tract and cystoscopy is recommended based on further risk calculation.

AVMs are fast-flow vascular malformations that comprise a complex network of primitive vessels directly connecting feeding arteries to draining veins (Uller et al., 2014). AVMs are usually congenital, though they may stay dormant during early childhood. They can vary greatly from patient to patient in angio-architectural features related to size, location, and venous drainage pattern (Osbun et al., 2017).

About 90% of AVMs are diagnosed by history and physical examination (Finn et al., 1983; Mulliken & Glowacki, 1982). Several methods of examination are utilized to detect the existence of AVMs. US is the first choice, with color Doppler examination showing fast flow and shunting. Magnetic resonance imaging (MRI) also is obtained to (a) confirm the diagnosis, (b) determine the extent of the lesion, and (c) plan treatment (Wu & Orbach, 2009). CT may be indicated if the AVM involves bone or some contraindications for MRI exist. Some researches indicate that CT arteriogram (CTA) is a superior choice as a preliminary diagnostic tool (Bilhim et al., 2014). The first reason is its high sensitivity and specificity. As a study reported, the sensitivity and specificity of CTA in detecting post-traumatic vascular lesions ranged from 90% to 100% and 98% to 100%, respectively (Soto et al., 2001). Second, CTA can discover clearly the location, size, and adjacent structure of the malformation. If the diagnosis is still unclear after US, CT, and MRI, it is essential to perform angiography. It is the gold standard in the diagnosis of AVM. Angiography is used not only to diagnose but also for therapy. Moreover, it can aid in determining the flow dynamics of the lesions. On angiography, AVM is mainly characterized by tortuous, dilated arteries with arteriovenous shunting and enlarged draining veins (Wu & Orbach, 2009). In this case, the patient's AVM of internal pudendal artery with hematuria was finally diagnosed by angiography.

The lesions of AVM are often diffuse, involving multiple tissue planes and important structures, and the possibility of cure is rare (Richter et al., 2007). The goal of treatment usually is to control the malformation. Intervention focuses on alleviating symptoms (e.g., bleeding, pain, ulceration), preserving vital functions (e.g., vision, mastication), and improving a deformity. The management includes embolization, resection, or a combination of the two. Resection may provide the best chance for long-term control, but the re-expansion rate is high and extirpation may cause a worse deformity. Embolization is not a curative method with the lesion's final re-expansion after embolization. Embolization is the most commonly method used preoperatively to reduce blood loss during resection or for a palliation of unresectable lesions (Greene & Orbach, 2011; Liu et al., 2010; Zaki Ghali et al., 2019).

In the present case, the treatment for AVM is embolization. Superselective arterial embolization of the internal pudendal artery is an accurate diagnostic tool as well as an elective and safe treatment for the control of bleeding. Embolic materials available for embolization include coils, ethanol, balloons, gelfoam particles and powder, and autologous clots (Po et al., 2012). Absorbable gelfoam particles, compared with permanent materials such as polyvinyl alcohol particles and trisacryl gelatin microspheres, are preferred because they are temporary embolic materials and without many complications. The complications of embolization for pelvic arteries include infection, necrosis of muscle or penis, nerve injury, bladder or ureteral infarction, bleeding or hemorrhage, bowel infarction, thigh or buttock claudication, and impotence (Vaidya et al., 2018). In this case, gelatin sponge and natural silk segments were used in order to preserve the normal erectile function of the patient. A very important reason for recurrence without necrosis of penis was the use of gelatin sponge the first time, which is the most common temporary embolization material. Thus, the second time the doctors chose gelatin sponge plus natural silk segments as the embolic material and infused slowly. The aim was

to give sufficient time for the formation of collateral circulation. It was the most important reason that no necrosis of penis occurred in 10 years after the treatment.

In conclusion, an AVM of the internal pudendal artery is a rare cause of recurrent painless gross hematuria. In clinical work, if common causes of unexplained hematuria occur, such as infection, trauma, or stone, they deserve high attention and a high index of suspicion is necessary for abnormal vascular issues, especially AVM. Imaging detections such as Doppler US, CTA, and angiography should be performed in such patients to reach an accurate diagnosis. It is important to protect functions of penis, while choosing the treatment of AVM of the internal pudendal artery. The selection of embolization materials and the process of operation also should be carefully handled.

Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Ethical statement

Ethics approval was not required for this study.

Patient consent

The patient of this case study consented to the publication of his information.

ORCID iD

Shixiang Qiu https://orcid.org/0000-0002-9754-1369

References

Ark, J. T., Alvarez, J. R., Koyama, T., Bassett, J. C., Blot, W. J., & Mumma, M. T. (2017). Variation in the diagnostic evaluation among persons with Hematuria: Influence of gender, race and risk Factors for Bladder cancer. *The Journal of Urology*, *198*(5), 1033–1038.

Bilhim, T., Pereira, J. A., Fernandes, L., Rio Tinto, H., & Pisco, J. M. (2014). Angiographic anatomy of the male pelvic arteries. AJR. *American Journal of Roentgenology*, *203*(4), W373–W382.

Bin_ Dahman, H. A., & Aljabry, A. O. (2019). A case report of a young girl with recurrent hematuria: A missed diagnosis - renal nutcracker syndrome. *BMC Nephrology*, *20*(1), 349.

Bolenz, C., Schroppel, B., Eisenhardt, A., & Schmitz-Drager, B. J. (2018). The investigation of Hematuria. *Deutsches Ärzteblatt International*, *115*(48), 801–807.

Celtikci, P., Ergun, O., Tatar, I. G., Conkbayir, I., & Hekimoglu, B. (2014). Superselective arterial embolization of pseudoaneurysm and arteriovenous fistula caused by transurethral resection of the prostate. *Polish Journal of Radiology*, *9*(79), 352–355.

Davis, R., Jones, J. S., Barocas, D. A., & Castle, E. P. (2012). Diagnosis, evaluation and follow-up of asymptomatic microhematuria (AMH) in adults: AUA guideline. *The Journal of Urology*, *188*(6), 2473–2481.

Finn, M. C., Glowacki, J., & Mulliken, J. B. (1983). Congenital vascular lesions: Clinical application of a new classification. *Journal of Pediatric Surgery*, *18*(6), 894–900.

Greene, A. K., & Orbach, D. B. (2011). Management of arteriovenous malformations. *Clinics in Plastic Surgery*, *38*(1): 95–106.

Jesinger, R. A., Thoreson, A. A., & Lamba, R. (2013). Abdominal and pelvic aneurysms and pseduoaneurysms: Imaging review with clinical, radiologic, and treatment correlation. *Radiographics*, *33*(3), E71–E96.

Joshua, A., Halpern, M., Bilal, C., & Hassan, G. (2017). Cost-effectiveness of common diagnostic approaches for evaluation of asymptomatic microscopic hematuria. *JAMA Internal Medicine*, *177*(6), 800–807.

Linder, B. J., Bass, E. J., Mostafid, H., & Boorjian, S. A. (2018). Guideline of guidelines: Asymptomatic microscopic haematuria. *BJU International*, *121*(2), 176–183.

Liu, A. S., Mulliken, J. B., Zurakowski, D., Fishman, S. J., & Greene, A. K. (2010). Extracranial arteriovenous malformations: Natural progression and recurrence after treatment. *Plastic and Reconstructive Surgery*, *125*(4), 1185–1194.

Mulliken, J. B., & Glowacki, J. (1982). Hemangiomas and vascular malformations in infants and children: A classification based on endothelial characteristics. *Plastic and Reconstructive Surgery*, *69*(3), 412–422.

Osbun, J. W., Reynolds, M. R., & Barrow, D. L. (2017). Arteriovenous malformations: Epidemiology, clinical presentation, and diagnostic evaluation. *Handbook of Clinical Neurology*, *143*(3), 25–29.

Papadakos, N., Wales, L., Hayes, K., Belli, AM., Loftus, I., & Ray, S. (2008). Post-traumatic plevic pseudoaneurysm and arterio-venous fistula: Combined endovascular and surgical approach. *European Journal of Vascular and Endovascular Surgery*, *36*(2), 164–166.

Peterson, L. M., & Reed, H. S. (2019). Hematuria. *Primary Care: Clinics in Office Practice*, *46*(2), 265–273.

Po, L. K., Simons, M. E., & Levinsky, E. S. (2012). Concealed postpartum hemorrhage treated with transcatheter arterial embolization. *Obstetrics and Gynecology*, *120*(2), 461–464.

Richter, G. T., North, P. E., James, C. A., Waner, M., & Buckmiller, L. M. (2007). Arteriovenous malformations of the tongue: A spectrum of disease. *Laryngoscope*, *1*(17), 328–335.

Schmitz-Drager, B. J., Kuckuck, E. C., Zuiverloon, T. C., Zwarthoff, E. C., Saltzman, A., & Srivastava, A. (2016). Microhematuria assessment an IBCN consensus-Based upon a critical review of current guidelines. *Urologic Oncology*, *34*(10), 437–451.

Soto, J. A., Munera, F., Morales, C., Lopera, J. E., Holguin, D., & Guarin, O. (2001). Focal arterial injuries of the proximal extremities: Helical CT arteriography as the initial method of diagnosis. *Radiology*, *218*(1), 188–194.

Uller, W., Alomari, A. I., & Richter, G. T. (2014). Arteriovenous malformations. *Seminars in Pediatric Surgery*, *23*(4), 203–207.

Vaidya, R., Waldron, J., Scott, A., & Nasr, K. (2018). Angiography and embolization in the management of bleeding pelvic fractures. *The Journal of the American Academy of Orthopaedic Surgeons*, *26*(4), e68–e76.

Valdez Vargas, A. D., Pérez Abarca, V. M., Maldonado Valadez, R. E., Badillo Santoyo, M. A., Sánchez López, H. M., & Manzo Pérez, B. O. (2018) Idiopathic renal arteriovenous fistula, a rare cause of severe hematuria: Case report. *Urology Case Reports*, *19*, 23–24.

Wu, I. C., & Orbach, D. B. (2009). Neurointerventional management of high-flow vascular malformations of the head and neck. *Neuroimaging Clinics of North America*, *19*, 219–240.

Zaki Ghali, G., Zaki Ghali, M. G., & Zaki Ghali, E. (2019). Transvenous embolization of arteriovenous malformations. *Clinical Neurology and Neurosurgery*, *178*, 70–76.